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Dear Editor

Anti-Leukotriene Therapy may be Effective for the Treatment of Salivary Eosinophilic Inflammation

Fibrinous sialodochitis is a very rare disease, and its most common symtoms include swelling of paroitid gland and excretion of sticky mucous plugs containing numerous eosinophils from Stensen's duct. Fibrinous sialodochitis has been believed to involve allergic response in the salivary duct and parotid glands. Unfortunately, the effective medical treatment except for the administration of glucocorticoid was not known. We experienced a case of fibrinous sialodochitis who was successfully treated with montelukast as follows:

On January 7, 2007, a 51-year-old woman presented to our hospital with a 3-year history of recurring bilateral parotid gland swelling and parotid pain during and after meals over 6 to 48 hours at least once per week. Previously, she visited two other ENT clinics for these symptoms. On questioning, the previous medication with antihistamines had no effect on these symptoms, while oral administration of prednisolone (10-20 mg/day) for one or two days palliated her parotid symptoms. She came to our hospital, expecting to receive the effective medication other than systemic steroids. She also complained of allergic rhinitis. Physical examination showed the nasal mucous membrane to be pale and edematous, the turbinates hypertrophied. An intraoral examination showed neither dryness nor atrophy of the oral mucous membrane. The blood examination except for the percentage of peripheral eosinophils (10.4%) and total IgE (295 kU/L) was within normal limits. Specific IgE levels of major inhaled allergens were also within normal limits. Examination for autoantibodies, such as the antinuclear antibody, SS-A and SS-B antibody, were all negative. Hansel staining showed that the mucous plugs from her Stensen's duct contained numerous eosinophils. Nasal smears also showed marked eosinophilia. A diagnosis of fibrinous sialodochitis complicated nonallergic rhinitis with eosinophilia syndrome was made. We started to the treatment of disease with an anti-allergic agent, suplatast tosilate (300 mg/day). However, this medicine was without any effect for her parotid symptoms. Then, medication was changed to an anti-leukotriene therapy with montelukast (10 mg/day). She recognized the initial objective improvement from two weeks after the start of the treatment. The frequency of cheeck swelling began to be lessened, and peripheral eosinophil levels were gradually decreased (Fig. 1). In adittion, there has been no exacerbation of her nasal symptoms. Athough we have proposed to reduce the dose of montelukast, she hopes to continue the medication with the same doses. To date these has been no adverse effects in this case.

Because the present case also complained of allergic rhinitis, we tried to use the cysteinyl leukotriene

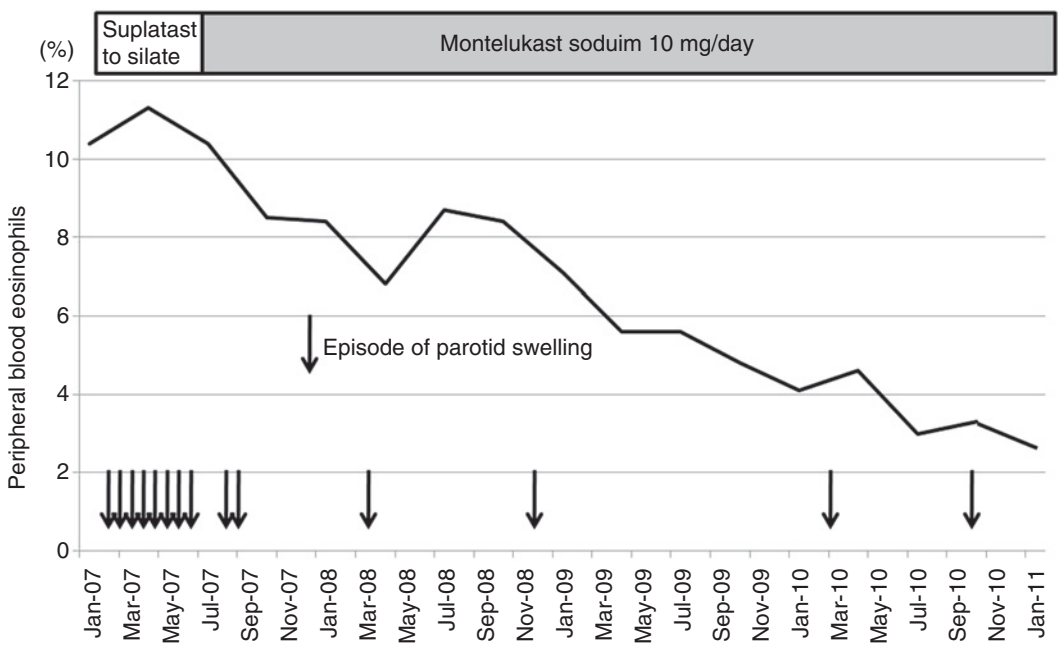

Fig. 1 Time course of clinical symptoms and the levels of eosinophils (%) in peripheral blood. Each arrow indicated the episode of cheeck swelling.

antagonist in spite of no previous report concerning the efficacy of cysteinyl leukotriene antagonists on fibrinous sialodochitis. In vitro, it has beeen reported that LTD4 extended the survival of eosinophils and CysLT1 receptor antagonist caused the apoptosis of eosinophils.[1] In previous clinical radomized trial, daily treatment with montelukast for 4 weeks reduced sputum and blood eosinophils and improved clinical asthma control with placebo.[2] So, CysLT1 receptor antagonists have been thought to have the benefits for eosinophilic inflammation. The clinical efficacy of montlelukast on the present case suggests that cysteinyl leukotrienes may play some important roles on fibrinous sialodochitis. Recently, higher concentrations of cysteinyl leukotrienes in saliva from asthmatic patients has been reported.[3] In order to confirm the roles of cysteinyl leukotrienes on fibrinous sialodochitis, the quantitative measurement of salivary cyslteinyl leukotrienes will be needed.

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